

available at <http://ajws.elsevier.com/tcmj>

Tzu Chi Medical Journal



Case Report

Tuberculosis Infection Within a Warthin's Tumor of the Parotid Gland

Yu-Hsuan Wen¹, Pei-Rong Chen^{1,2}, Hung-Pin Wu^{2,3,4*}

¹Department of Otolaryngology, Buddhist Tzu Chi General Hospital, Hualien, Taiwan

²Department of Medicine, College of Medicine, Tzu Chi University, Hualien, Taiwan

³Department of Otolaryngology, Buddhist Tzu Chi General Hospital Taipei Branch, Taipei, Taiwan

⁴Institute of Occupational Medicine and Industrial Hygiene, National Taiwan University, Taipei, Taiwan

Article info

Article history:

Received: December 18, 2007

Revised: January 2, 2008

Accepted: January 28, 2008

Keywords:

Parotid gland

Tuberculosis

Warthin's tumor

Abstract

The parotid gland is an extremely rare site for extrapulmonary tuberculosis, even in countries where the disease is widespread. Clinically, tuberculosis of the parotid gland is indistinguishable from other parotid tumors because it frequently presents as a neoplasm. Most patients undergo parotidectomy and the tissue diagnosis changes the clinical impression. We report an 81-year-old man who had a 3×3 cm infra-auricular mass for half a year. Pathology demonstrated Warthin's tumor and granulomatous stroma. Acid-fast stain was positive. Pulmonary tuberculosis and tuberculosis within the Warthin's tumor were identified. The patient was treated with antituberculous medication and was disease-free after 12 months of follow up. (*Tzu Chi Med J* 2008;20(4):332–334)

*Corresponding author. Department of Otolaryngology, Buddhist Tzu Chi General Hospital, Taipei Branch, 289, Jianguo Road, Taipei, Taiwan.
E-mail address: hungpin_wu@yahoo.com.tw

1. Introduction

Warthin's tumor (papillary cystadenoma lymphomatosum) is the second most common neoplasm of the parotid gland and has been reported to account for up to 6–30% of parotid neoplasms (1). However, parotid tuberculosis is rare, and preoperative diagnosis is difficult (2,3). In a review of the English literature, only a few cases of tuberculosis within a Warthin's tumor have been reported (4). We report a case of Warthin's tumor of the parotid gland with tuberculosis within it.

2. Case report

An 81-year-old man presented with a 6-month history of a progressively enlarging painless swelling in the

right infra-auricular region. He had no other symptoms or complaints, such as facial weakness, fever or prolonged cough. He had a medical history of hypertension and diabetes mellitus. All laboratory tests were within the normal ranges. However, chest X-ray before surgery revealed multiple nodular lesions in bilateral lung fields (Fig. 1). Computed tomography revealed an ovoid well-enhanced mass with multiple central cystic changes over the right parotid gland (Fig. 2).

Under the diagnosis of right parotid Warthin's tumor, a superficial parotidectomy was carried out. The gross specimen measured 3.2×2.9×2.8 cm. Pathological examination revealed proliferative oncocytic columnar epithelium and markedly lymphocytic infiltration within the stroma. About half of the mass was occupied by chronic granulomatous inflammation with focal necrosis and multinucleated giant cells (some were of



Fig. 1 — Multiple nodular lesions in bilateral lung fields.



Fig. 2 — Computed tomography shows an ovoid well-enhanced mass with multiple central cystic changes over the right parotid gland.

the Langerhans' giant cell type) (Fig. 3). Acid-fast bacilli were seen on acid-fast stain (Fig. 4).

The patient was stable postoperatively and was discharged 7 days after surgery. Cultures of both the parotid specimen and sputum later revealed *Mycobacterium tuberculosis*. After discharge, the patient received antituberculous treatment with ethambutol and Rifater (rifampicin, isoniazid and pyrazinamide; Sanofi-Aventis, Paris, France) in the outpatient clinic. After 12 months of follow-up, the sputum culture was

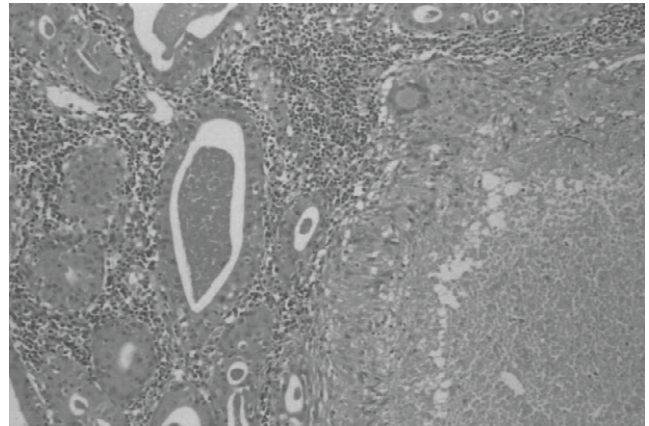


Fig. 3 — Warthin's tumor is seen on the left and chronic granulomatous inflammation with focal necrosis and multinucleated giant cells on the right (40×).

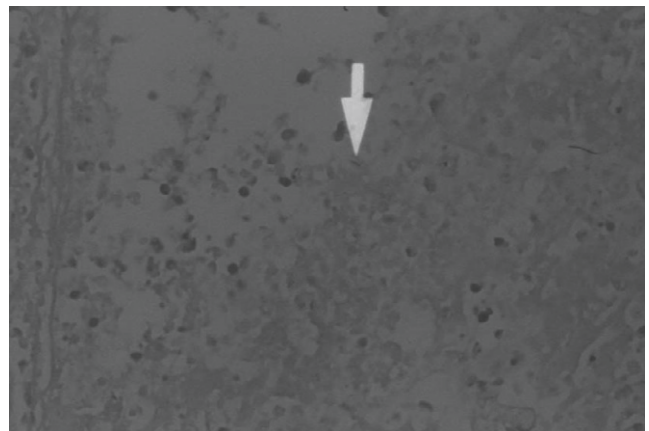


Fig. 4 — Acid-fast bacilli (arrow) were found in tuberculosis granuloma (100×).

negative for tuberculosis, and chest radiography showed no evidence of active tuberculosis infection.

3. Discussion

Warthin's tumor typically presents as a slowly growing mass in the tail of the parotid gland. The majority of cases occur among patients aged 50–60 years. A superficial parotidectomy is usually the treatment of choice (1). According to the clinical manifestations and imaging findings in our patient, Warthin's tumor was diagnosed before surgery. Pathological examination of the specimen showed that in addition to proliferative oncocytic columnar epithelium and marked lymphocytic infiltration within the stroma (which are typical findings of Warthin's tumor), half of the mass was occupied by chronic granulomatous

inflammation with focal necrosis and multinucleated giant cells. Acid-fast bacilli were seen on acid-fast stain. Cultures of both the mass and sputum revealed *Mycobacterium tuberculosis*.

In contrast to Warthin's tumor, tuberculosis is a rare cause of parotid swelling. The diagnosis is often difficult to make based on clinical manifestations only (2,3,5). It is also frequently impossible to distinguish tuberculosis from parotid gland neoplasms by clinical or sonographic investigation, even with fine needle aspiration cytology (6). Most cases are diagnosed after histopathologic examination (7,8). Parotid tuberculosis can be part of systemic mycobacterial disease or the primary focus (8). Tuberculous lymphadenitis is the most common extrapulmonary form of tuberculosis, and the cervical lymph nodes in and around the salivary glands are most frequently involved (5).

Parotid tuberculosis may develop in two different ways. Primary salivary tuberculosis may begin as an infection from the tonsils or teeth, or infect the gland via the gland's ducts or their associated lymph nodes via lymphatic drainage (5). In secondary salivary tuberculosis, the gland may be involved via hematogenous or lymphatic spread from the lungs (5). Tuberculosis is normally confined to the intraglandular and periglandular lymph nodes in the major glands, and invasion of the parenchyma is usually secondary spread from the nodes themselves (7). Because the chest radiographs in this case revealed multiple nodular lesions in bilateral lung fields, pulmonary tuberculosis was suspected and secondary infection from the lung to the parotid gland was favored. There was no obvious evidence to suggest that tuberculosis occurred before or after Warthin's tumor. The association of Warthin's tumor with parotid tuberculosis could have been a coincidence.

Primary salivary tuberculosis involves the parotid gland more frequently than the other salivary glands. But secondary salivary tuberculosis may also occur in the parotid gland, although it involves the submandibular or sublingual glands more frequently and is associated with pulmonary tuberculosis. Clinically, localized tuberculosis involves the parotid gland, while the submandibular gland is affected in systemic tuberculosis disease (8). Tuberculosis should be considered in the differential diagnosis of patients who present with a parotid tumor, especially in those with

a past history of tuberculosis infection or evidence of tuberculosis elsewhere. Extrapulmonary mycobacterial infection may occur in many organs as part of systemic processes in the absence of clinical disease in the lungs. O'Connell et al described six patients with mycobacterial infection of the parotid gland, none of which had lung lesions. It usually presented as a slowly growing mass, clinically indistinguishable from a parotid tumor (9).

Tuberculosis infection within Warthin's tumor is seldom seen. The treatment for tuberculosis should be antituberculous chemotherapy, not surgery (2). In contrast to tuberculosis infection, surgical intervention is indicated for an enlarged Warthin's tumor. Mass excision should be followed by a complete medical treatment course, regardless of whether or not there is lung involvement. In patients with suspected mycobacterial infection, further consideration of extrapulmonary tuberculosis infection to organs or tumor should be emphasized.

References

1. Yoo GH, Eisele DW, Askin FB, Driben JS, John ME. Warthin's tumor: a 40-year experience at The Johns Hopkins Hospital. *Laryngoscope* 1994;104:799-803.
2. Singh B, Maharaj TJ. Tuberculosis of the parotid gland: clinically indistinguishable from a neoplasm. *J Laryngol Otol* 1992;106:929-31.
3. Hunter DC, Thomas JM. Tuberculosis in the parotid region. *Br J Surg* 1993;80:1008.
4. Yamada N, Uchinuma E, Nanjo A, et al. A tuberculoma in an adenolymphoma arising in the parotid glands: a case report. *Kitasato Med* 1996;26:99-101.
5. Hamdan AL, Hadi U, Shabb N. Tuberculous parotitis: a forgotten entity. *Otolaryngol Head Neck Surg* 2002;126:581-2.
6. Franzen A, Franzen CK, Koegel K. Tuberculosis of the parotid gland: a rare differential diagnosis of parotid tumor. *Laryngorhinootologie* 1997;76:308-11.
7. Holmes S, Gleeson MJ, Cawson RA. Mycobacterial disease of the parotid gland. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 2000;90:292-8.
8. Süog'lu Y, Erdamar B, Çölhan L, Katircioglu OS, Cevikbas U. Pathology in focus tuberculosis of the parotid gland. *J Laryngol Otol* 1998;112:588-91.
9. O'Connell JE, George MK, Speculand B, Pahor AL. Mycobacterial infection of the parotid gland: an unusual cause of parotid swelling. *J Laryngol Otol* 1993;107:561-4.